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## CONGENITAL ABSENCE OF LEFT PULMONARY ARTERY

BY

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The congenital absence of a pulmonary artery is a rare condition, but one which has been recognized for many years. The first case was reported by Fraentzel (1868), this being discovered at necropsy, and, later, Döring (1914), Müller (1927), Ambrus (1936), Miller (1937), and Thomas (1941) all recorded cases of absent pulmonary artery found after death.

Subsequently, further cases have been reported, largely occurring in cyanotic heart disease, the absent pulmonary artery being found at operation. Blalock (1947, 1948) refers to nine such patients occurring in a series of 610 cases of cyanotic heart disease. Findlay and Maier (1951) and Jew and Gross (1952) reported further examples of this condition found at operation.

After the introduction of angiocardiology the diagnosis of this condition became possible without operation, and from 1952 onwards a number of cases of an absent pulmonary artery were described. Madoff *et al.* (1952), Steinberg *et al.* (1953), Madoff (1954), Wyman (1954), and Flynn *et al.* (1954) all described cases of an absent pulmonary artery in patients with a normal heart, diagnosed by angiocardiology. During this same period Nadas *et al.* (1953), McKim and Wiglesworth (1954), Wyman (1954), Campbell (1954), and Maier (1954) reported cases of an absent pulmonary artery associated with congenital heart disease, and some of the cyanotic type. Finally, Read (1954) reported a case during the discussion on Maier's paper in which there was an absent pulmonary artery associated with hypoplasia of the lung; and Waterman (1954), also during the discussion of Maier's paper, reported two cases of very small right pulmonary arteries.

It would appear from the literature that the absence of a pulmonary artery may occur on either side, but is slightly more common on the left than on the right, and

is often associated with congenital heart disease, particularly of the cyanotic type. In those recorded cases where there is no evidence of heart disease, the absence of the right pulmonary artery appears to be more frequent than the absence of the left. The defect occurs more commonly in females than in males. This variation of an absent left pulmonary artery with congenital heart disease and an absent right pulmonary artery with a normal heart has been noted by Emanuel and Pattinson (1956).

The following case is reported in view of the rarity of the condition and its radiological interest.

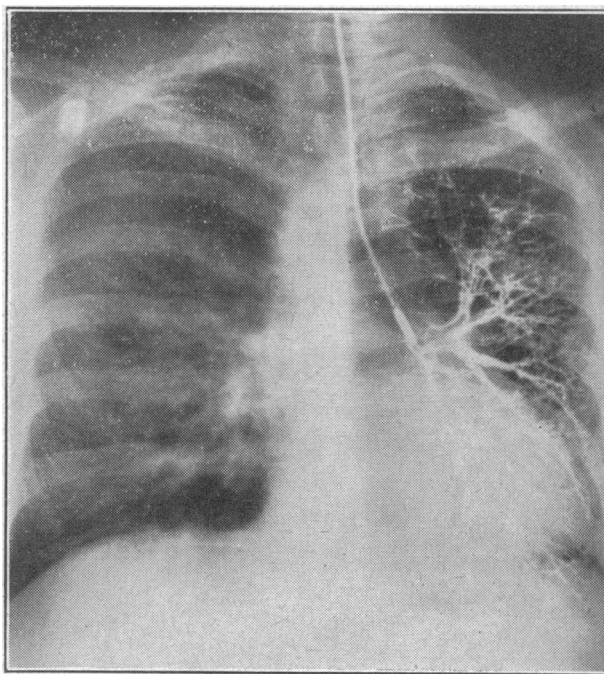


FIG. 1.—Normal left bronchogram.

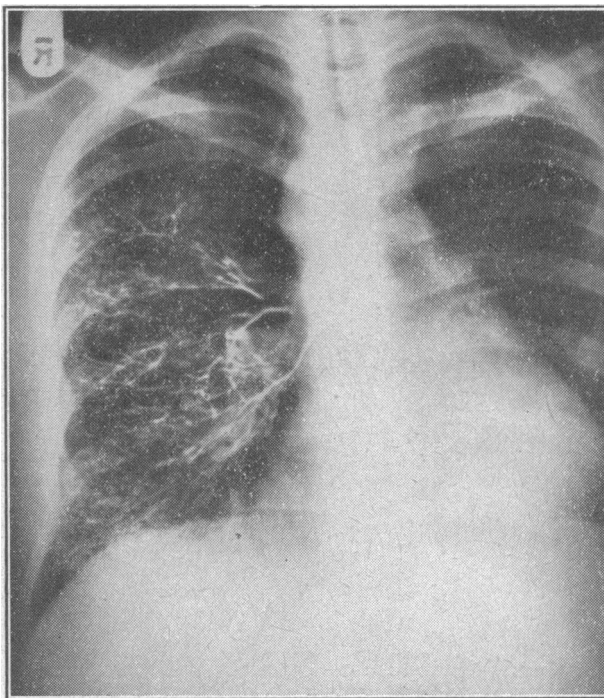


FIG. 2.—Normal right bronchogram.

## Case Report

A man aged 30 was referred to one of us (J.S.) for investigation following a routine mass miniature x-ray examination. He had no symptoms of any sort, and on clinical examination appeared to be a healthy man. There was no cyanosis, and no abnormal physical signs were found in the respiratory or cardiovascular systems. A radiograph of the chest showed the left lung to be smaller than the right, and there was a moderate degree of displacement of the heart and mediastinum to the left. Owing to this displacement, the left hilum was hidden, but at the right hilum a pulmonary artery shadow rather larger than normal was visible. The right lung showed a slight increase in translucency.

On fluoroscopy both domes of the diaphragm moved equally and well, and there was no evidence of collapse of any part of the left lung. Congenital absence of a segment or lobe of the left lung was suspected. Bronchograms, however, showed a perfectly normal bronchial tree on both sides (Figs. 1 and 2). It was also observed that the aortic arch and the upper part of the descending aorta lay on the right side of the trachea, and that owing to the mediastinal displacement their shadow was superimposed on that of the spine. The presence of a right-sided aortic arch was subsequently confirmed by a barium swallow examination. There was no evidence of a retro-oesophageal vessel or diverticulum. On the bronchogram films the large right pulmonary artery was seen, but no shadow of a left pulmonary artery could be detected at the left hilum. A provisional diagnosis of congenital absence of the left pulmonary artery was therefore made. An E.C.G. revealed no abnormality at rest or at exercise.

The angiocardigram showed a normal main pulmonary trunk ascending from the right ventricle. This vessel did not bifurcate, but continued in an even curve to the right to become continuous with the large right pulmonary artery. There was no evidence of a normal left pulmonary artery (Fig. 3), the only pulmonary vessel supplying the left lung being a tiny vessel 2 mm. in diameter passing down to the lower lobe. Subsequent films showed the right-sided aortic arch with the descending aorta crossing to the left of the midline in the lower third of the thorax. The virtually complete absence of a pulmonary supply to the left lung

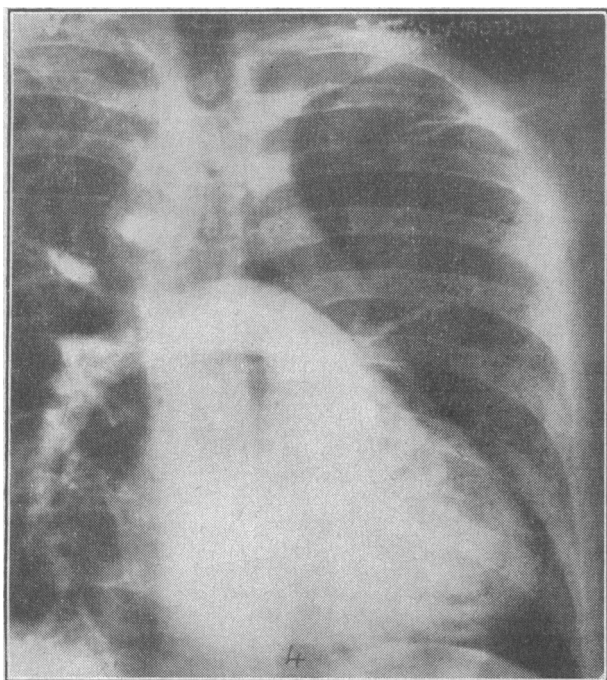


FIG. 3.—Angiocardiogram 4 seconds after injection of dye, showing absence of left pulmonary artery, except for a minute branch to left lung.

suggested that the latter served no useful function. Bronchspirometry showed that the ventilation of each lung was approximately equal, the vital capacity of the right lung being 2,800 ml., and of the left 2,350 ml. The oxygen uptake on the right side was 390 ml. a minute, while on the left side no oxygen was absorbed (Fig. 4).

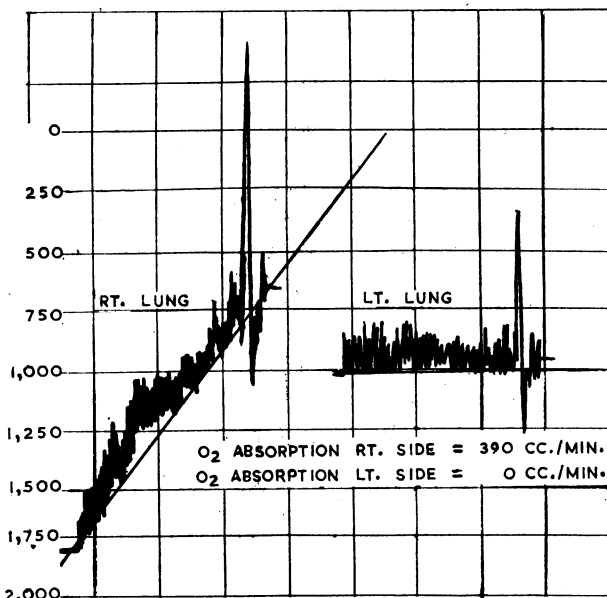


FIG. 4.—Bronchspirometry tracing, showing oxygen absorption of 390 ml./min. by the right lung and no oxygen absorption by the left.

## Discussion

Bronchspirometry studies were carried out by Madoff *et al.* (1952) and in the further case reported by Madoff (1954). In one case, that of a girl aged 14 years, bronchspirometry showed that the oxygen uptake on the right side was 7% and on the left 93%, but when air was substituted for the oxygen there was apparently no uptake of oxygen on the right side. They also did bronchspirometry studies on a man aged 23 who had no right pulmonary artery, and in this case the oxygen uptake was 16% on the right side and 84% on the left. When this patient died a few months later it was found that he had a persistent ductus arteriosus and an infantile type of coarctation of the aorta, but no anomalous artery to the right lung, though the left pulmonary artery did not appear to be enlarged. The uptake of oxygen in the first case is difficult to understand unless it was via the bronchial arteries; and, if this was so, it would possibly explain why, in the second case, the uptake from the right side, with an absent right pulmonary artery, was higher, because this patient had a persistent ductus and therefore a shunt, which resulted in some mixing of the blood, a lowered oxygen saturation, and consequently an increased uptake from the bronchial artery. Our results of bronchspirometry, even with oxygen, did not show any uptake on the affected side.

Absence of one of the pulmonary arteries in itself produces few or no symptoms. In cases where there is no significant cardiac defect the patient may be asymptomatic or there may be slight dyspnoea, cough, or occasional haemoptysis from the bronchial arterial supply to the affected lung.

In cases of absence of a pulmonary artery the diagnosis can be suspected from the plain radiographs. On the affected side the lung is usually smaller than the other one, with small intrapulmonary vascular shadows. There may be poor development of the hemithorax and poor movement of the diaphragm on that side. The contralateral lung is often enlarged, showing increased radiolucence, and may permeate across the midline. The mediastinum is often

displaced towards the affected side. The presence of a large pulmonary artery, which may even be aneurysmal, on the contralateral side and the absence of the shadow of a normal pulmonary artery at the hilum on the affected side are important signs. The latter feature may be difficult to appreciate on routine films, for the displaced heart may obscure the hilum. Penetrated films and tomograms of the hila are therefore of value. Bronchograms usually show a normal bronchial tree, as in the patient reported here, but in a few of the reported cases the affected lung has been small, in one case unilobar and in others agenetic. Confirmation of the absence of one of the pulmonary arteries can be obtained by angiocardiology.

### Summary

A case of congenital absence of the left pulmonary artery occurring asymptotically in a man of 30, associated with a right-sided aorta, is recorded. The literature is reviewed.

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## PLASMA ACTIVITY LEVELS IN RADIO- IODINE TESTS OF THYROID FUNCTION

BY

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In an investigation carried out at Sheffield with radio-active iodine (Goodwin *et al.*, 1951 ; Ansell *et al.*, 1953 ; Wayne, 1954) it was found that the most useful single index of thyroid function was the estimation of the 48-hour protein-bound plasma activity, and that measurement of the 48-hour total plasma activity also gave valuable if less definite diagnostic data. It was decided to adopt the latter estimation as a routine test of thyroid function in this hospital as it seemed more suited to the facilities available. The Sheffield workers drew their patients from an iodine-deficient area, and, so far as we are aware, there are very few published results of plasma-activity estimations from other parts of Great Britain ; thus the results described in this paper may be of some interest, as the patients were all drawn from South-east England.

In addition to the 48-hour total plasma activity, the following indices were determined in a limited number of cases: the ratio of total plasma activity at 48 hours and 2 hours (Blondal, 1952 ; Fellingner *et al.*, 1953), the "T" index (Fraser *et al.*, 1953) based on urinary excretion of <sup>131</sup>I, and the 0-24 hours urinary excretion. The last index affords a further comparison between our results and those of the Sheffield workers.

### Material

Eighty patients were investigated, their clinical condition being assessed in terms of the classification used by Fraser *et al.* (1953). They were divided into three groups: (1) Ten patients with marked hyperthyroidism—five with severe toxic goitre and five with severe exophthalmic Graves's disease. (2) An intermediate group of 20 patients—11 with mild toxic goitre and 9 with mild exophthalmic Graves's disease. (3) A group of 50 patients sent to hospital with suspected hyperthyroidism but subsequently judged on clinical grounds to have normal thyroid function.

Patients with mild hyperthyroidism or normal thyroid function form the majority of our series, as we felt that

TABLE I

	B.M.R. (%)	24-hour Urinary Excretion (%)	"T" Index	48-hour Total Plasma Activity (% Dose/ litre)	48: 2- hour Ratio
<i>Results of Tests in Normal Group (50 Cases)</i>					
Range ..	-17 to +74	21.9-70.5	1.55-20.6	0-0.57	0-0.28
No. in range	16	14	12	44	16
Mean ..	14.8	47.3	6.46	0.15	0.05
S.D. ..	18	15.2	5.72	0.14	0.06
<i>Results of Tests in Intermediate Group (20 Cases)</i>					
Range ..	-1 to +90	6.2-61.0	1.68-167	0-1.88	0-2.98
No. in range	13	11	10	18	11
Mean ..	36	26.2	27.76	0.69	0.69
S.D. ..	27	16.2	48.52	0.57	0.83
<i>Results of Tests in Toxic Group (10 Cases)</i>					
Range ..	+37 to +63	3.2-17.9	11.1-37.7	0.72-3.0	0.93-5.75
No. in range	4	5	5	10	4
Mean ..	47.8	11.5	26.8	2.06	2.93
S.D. ..	10.2	4.6	9.8	0.84	1.97

TABLE II.—Atypical Cases\*

Case No.	Sex	B.M.R. (%)	24-hour Urinary Excretion (%)	"T" Index	48-hour Total Plasma Activity (% Dose/ litre)	48: 2- hour Ratio	Diagnosis
16	F	+34	9.7	49.2	1.31	0.88	Mild exophthalmic Graves's disease
18	F	+35	16.5	8.54	1.88	1.18	Mild toxic goitre
24	F	+65	6.2	16.7	1.4	2.98	" "

\* These cases have been placed in the intermediate group.

TABLE III

	B.M.R. (%)	24-hour Urinary Excretion (%)	"T" Index	48-hour Total Plasma Activity (% Dose/ litre)	48: 2- hour Ratio
<i>Intermediate Group—Subgroup A (10 Cases*), Mild Toxic Goitre</i>					
Range ..	+2 to +81	8.8-39.7	4.22-25.5	0.05-1.88	0.01-1.18
No. in range	5	5	4	9	5
Mean ..	36	23.0	11.24	0.70	0.45
S.D. ..	26	13.1	8.10	0.79	0.44
<i>Intermediate Group—Subgroup B (8 Cases†), Mild Exophthalmic Graves's Disease</i>					
Range ..	-1 to +90	23.3-61.0	1.68-6.05	0-1.34	0-0.32
No. in range	5	4	4	8	4
Mean ..	34	39.4	3.36	0.54	0.14
S.D. ..	31.7	14.7	1.63	0.48	0.12

\* Atypical Case 24 omitted. † Atypical Case 16 omitted.